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CASE REPORT

Chronic osteomyelitis with proliferative periostitis in the lower jaw

Yen-Ching Chang [a,e], Yi-Shing Shieh [b,e], Shiao-Pieng Lee [a,e], Yi-Jan Hsia [a,e], Chih-Kung Lin [c], Shin Nieh [c], Huey-Kang Sytwu [d], Yuan-Wu Chen [a,e*]

[a] *Department of Oral and Maxillofacial Surgery, Tri-Service General Hospital, Taipei, Taiwan*
[b] *Department of Family Dentistry and Oral Diagnosis, Tri-Service General Hospital, Taipei, Taiwan*
[c] *Department of Pathology, Tri-Service General Hospital, Taipei, Taiwan*
[d] *Department of Microbiology and Immunology, National Defense Medical Center, Taipei, Taiwan*
[e] *School of Dentistry, National Defense Medical Center, Taipei, Taiwan*

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Abstract Chronic osteomyelitis with proliferative periostitis (Garré's sclerosing osteomyelitis) is a distinctive type of chronic osteomyelitis that mainly affects children and young adults. Here we report on a 9-year-old girl in whom the condition arose following a pulpoperiapical infection in a mandibular right primary secondary molar. Clinically, it manifested as a bony, hard, mildly tender swelling. Radiography revealed a pathognomonic patchy thickening with radiolucency and radiopacity. The dental inflammation and infection were eliminated and conservative therapy followed. The patient was otherwise asymptomatic. Remission of the disease process and reappearance of a normal-looking mandible was observed with computed tomography imaging, three-dimensional reconstruction and a bone scan at a 10-month follow up visit.

Introduction

Garré's sclerosing osteomyelitis was first described by Carl Garré in 1893 as irritation-induced focal thickening of the periosteum and cortical bone of the tibia.[1] It is a type of chronic osteomyelitis that primarily affects children and adolescents.[2] In the orthopedic literature, chronic osteomyelitis with proliferative periostitis (COPP) of the tibia is a well-known syndrome. Berger described the first cases of proliferative periostitis affecting the jaw bones. Reports of COPP in the body of the mandible are relatively common.[3] Most reported cases are unilateral, but one case involving all four quadrants of the jaw has been reported.

* Corresponding author. Number 325, Cheng-Kung Road, Section 2, Neihu 114, Taipei, Taiwan, ROC.
E-mail address: h6183@yahoo.com.tw (Y.-W. Chen).

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COPP is a more accurate description of the pathogenesis of this condition than Garré's osteomyelitis, as mild irritation and infection affect the jaw and lead to peripheral subperiosteal bone deposition.[4] This process, related to periosteal osteoblastic activity formation of subperiosteal bone, represents a periosteal reaction to inflammation. The histopathology of COPP is actually distinctive. The affected periosteum forms several layers of vital bone that are parallel to each other and to the surface of the affected bone. An intact cortex is present below the new bone formation.

COPP commonly occurs in young patients with a mean age of 13 years. Sporadic cases have been reported in patients in their 20s and in infants as young as 2 years. COPP is a non-suppurative inflammatory process. The most common provoking factors in the jaw region are a previous tooth extraction, tooth eruption, dental caries with associated periapical inflammation, periodontal infections, fractures and non-odontogenic infections. Most cases arise in the molar/premolar area of the mandible. Clinically, patients may present with a hard swelling of the jaw and facial asymmetry caused by this reactive process.

Radiographic examinations typically show bony laminations parallel to each other and to the cortical surface of the involved bone. Areas of small sequestra or osteolytic radiolucencies can be found within the new bone. Appropriate radiographic angulation can highlight a radiolucent zone of soft tissue between the original bony cortex and the newly-formed reactive bone. COPP presents a special radiographic feature, especially evident in computed tomography (CT) with three-dimensional reconstruction, with new periosteal proliferation located in successive layers parallel to the condensed cortical bone.[5] This phenomenon is also correlated with typical radiographic features such as an "onion skin" appearance. Other lesions that must be considered in differential diagnosis of COPP are Ewing's sarcoma, fibrous dysplasia, osteogenic sarcoma, infantile cortical hyperostosis, callus, exostosis, calcifying hematoma, and osteotomas.

In this case, a 9-year-old girl was evaluated based on her clinical, radiographic and histopathological features. Clearly, in lesions of this type, histopathology plays a major role in determining the final diagnosis and subsequent course of treatment.

Case report

A 9-year-old girl presented with extra-oral swelling in the right inferior border of the mandible. Extra-oral examination of the right side revealed a diffuse, mildly tender swelling, which was hard in consistency with no lymphadenopathy. The skin color was normal. An intraoral photograph showed a postextraction (85) wound in relation to area 44–46 (extracted by a general practitioner 20 days previously). Periodontal probing around the tooth revealed no deep pocketing. There was a functional opposing tooth. The patient's history revealed she had been seen by several practitioners and had been treated without resolution for an assumed dentoalveolar abscess with various courses of antibiotics.

At the patient's first presentation at our outpatient department, plain radiographs (Fig. 1A) and CT scans demonstrated enlargement of the affected mandible. They showed patchy areas of sclerosis and relative radiolucencies together with thickening of the overlying soft tissues (Fig. 1B and C). Increased tracer uptake in the region of the right mandible was evident on a bone scan; there was no significant scintigraphic activity except for the epiphyseal plates (Fig. 2A). A full blood count, serum levels of urea, electrolytes, calcium, and phosphate were within normal limits. A raised erythrocyte sedimentation rate (24 mm/h) and alkaline phosphatase (221 U/L) were consistent with the clinical examination. We made a provisional diagnosis of neoplastic bone, with inflammatory-infectious and fibro-osseous changes to the mandible.

Histopathology of a bone biopsy examination revealed a chronic nonspecific inflammatory lesion without any evidence of granulomatous inflammation; a microbiological culture was also negative. The biopsy exhibited trabeculae of bone with osteoblastic rimming and reversal lines (Fig. 2B). Based on the patient's clinical presentation, radiographic characteristics and histopathology, we diagnosed COPP. We administered a non-steroidal anti-inflammatory drug and the symptoms and signs subsided in 7 days. We closely followed this patient and at a 10-month follow-up visit, we found the patient to be asymptomatic with no signs (Fig. 3A and B). Occlusal, panoramic radiographs (Fig. 3C) and CT scans showed remodeling of the jaw (Fig. 4A). The previous increased tracer uptake in the region of the right mandible had returned to normal on a bone scan (Fig. 4B).

Discussion

The patient's symptoms and radiographic and clinical features were consistent with several different disease categories, including neoplastic, fibro-osseous and inflammatory-infectious diseases.

The lack of fever, limited response to antibiotic therapy, enlargement of the lesion, lack of suppuration and radiographic features of a loss of the mandibular border were consistent with a malignancy. The possible specific neoplasms included mesenchymal tumors such as an osteosarcoma, a chondrosarcoma, and Ewing's sarcoma. This variation in radiographic presentation is caused by the varied forms of osteosarcoma (lytic, sclerosing, or mixed).[6] Osteolytic mandibular lesions are typically bulky, with irregular radiolucency including both expansion and destruction of the cortical plate. For chondrosarcomas, a "sun-ray" and "onion skin" appearance have also been described.[7,8] For Ewing's sarcoma, onion skinning is also a typical radiographic feature.[9] However, the histopathology in this case revealed a chronic nonspecific inflammatory lesion without malignant changes (Fig. 2B).

A fibrous dysplasia needed to be considered because this type of lesion is frequently seen in the craniomaxillofacial region as a painful enlargement of the periosteal bone, and its characteristic radiographic features could be consistent with those seen in this patient. Although more frequently found in the maxilla, fibrous dysplasia can occur in the mandible of young patients as a focal enlargement.[10]

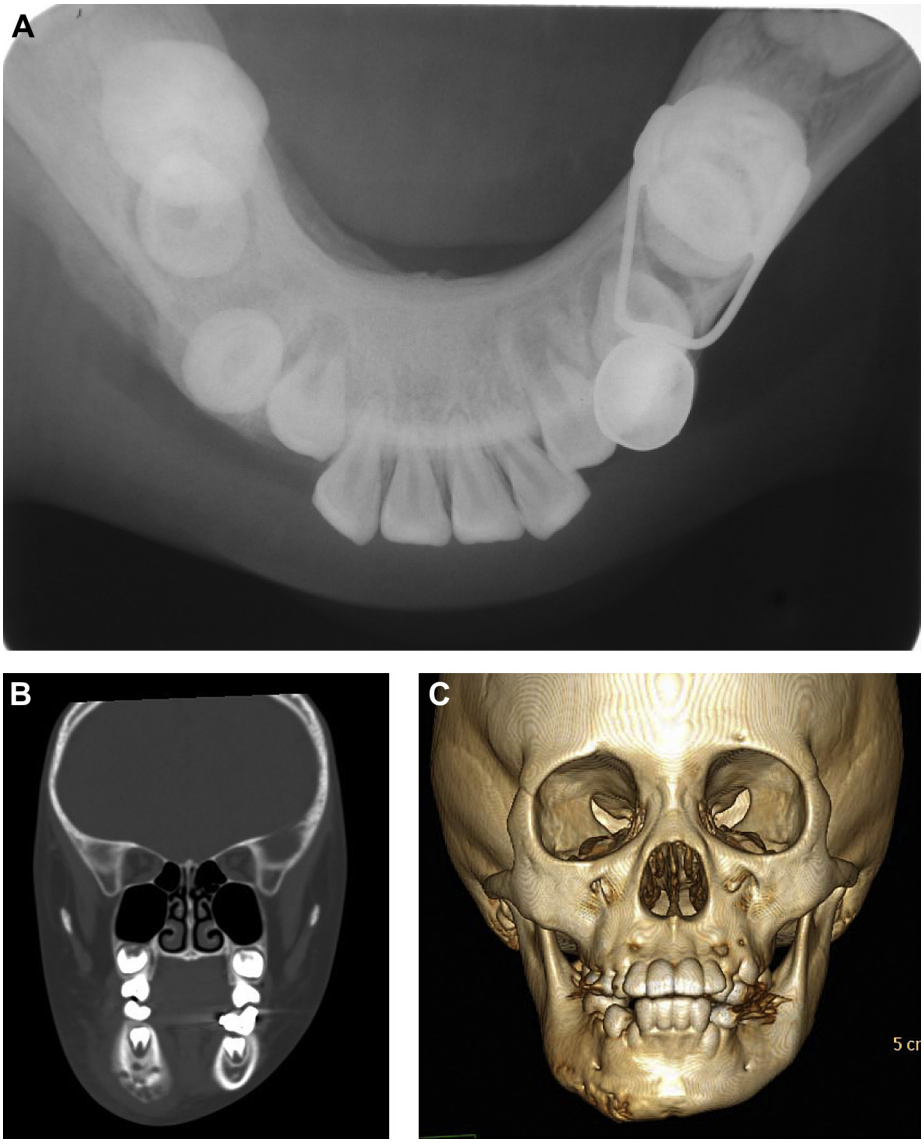

Figure 1 (A) An occlusal radiograph of the mandible showed the enhancement in peripheral subperiosteal bone on the right buccal and lingual sides. (B) Coronal section computed tomography of the bulging area demonstrated severe enhancement over the periosteum. (C) Coronal section three-dimensional computed tomography of the bulging area showed an additional irregular region of cortical bone extending from the parasymphysis to the right retromolar area.

Usually histopathological examination reveals a trans-differentiation from fibrous components to immature bone structures, without osteoblastic rimming, in the early process of fibrous dysplasia. In addition, the bony trabeculi in early fibrous dysplasia were much more irregular than those of the present case.

Osteomyelitis can occur in existing fibrous dysplasia or in benign fibro-osseous disease of the periodontal ligament following biopsy or surgical treatment. However, the microbiological culture was negative and histopathology revealed no fibrotic changes.

In this case, the inflammatory-infectious etiology could be narrowed to include osteomyelitis, specifically the neo-periostoses such as non-suppurative ossifying periostitis, periostitis ossificans or non-suppurative sclerosing osteomyelitis with proliferative periostitis (Garré's osteomyelitis). Conventional occlusal and panoramic radiography are generally appropriate for making a diagnosis in patients with COPP. One prior study[5] classified COPP into two types, based on whether the original contour of the mandible was preserved as seen by radiography. Type II involves loss of contour of the mandible and is also divided into two subtypes. Type II subtype 1 shows newly formed bony enlargement with resorption of the original cortex and osteolytic areas are usually visible. The Type II subtype 2 shows deformation with a homogeneously dense osteosclerotic bone that makes the original cortex discernible. This subtype occasionally includes duplication of newly formed periosteal bone on the outer aspect of the deformed mandible. Our case presented a newly formed bony enlargement with resorption of the original cortex and osteolytic areas, which defined it as Type II subtype 1 (Fig. 1B and C).

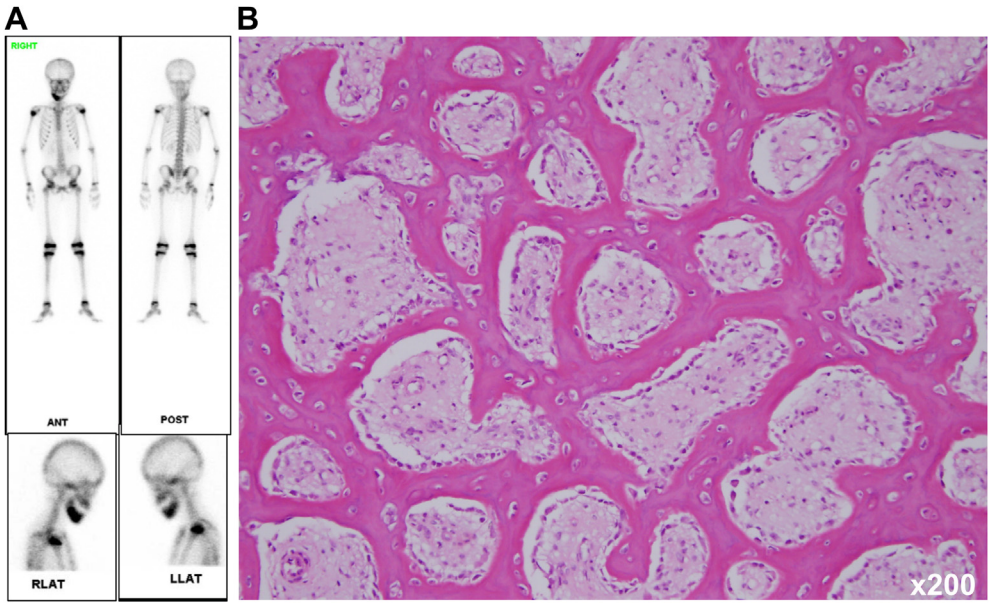

Figure 2 (A) Whole-body bone scans demonstrated intense accumulation of tracer in the right mandibular area and in epiphyseal plates at each end of a long bone. (B) A photomicrograph of the biopsy revealed the mixed woven bone, mature bone, osteoblastic rimming, reversal lines, mild fibrotic changes in the marrow space and mild inflammatory cellular infiltration, which is consistent with a bone repairing process after a chronic infection or wound (hematoxylin and eosin staining, ×200 magnification).

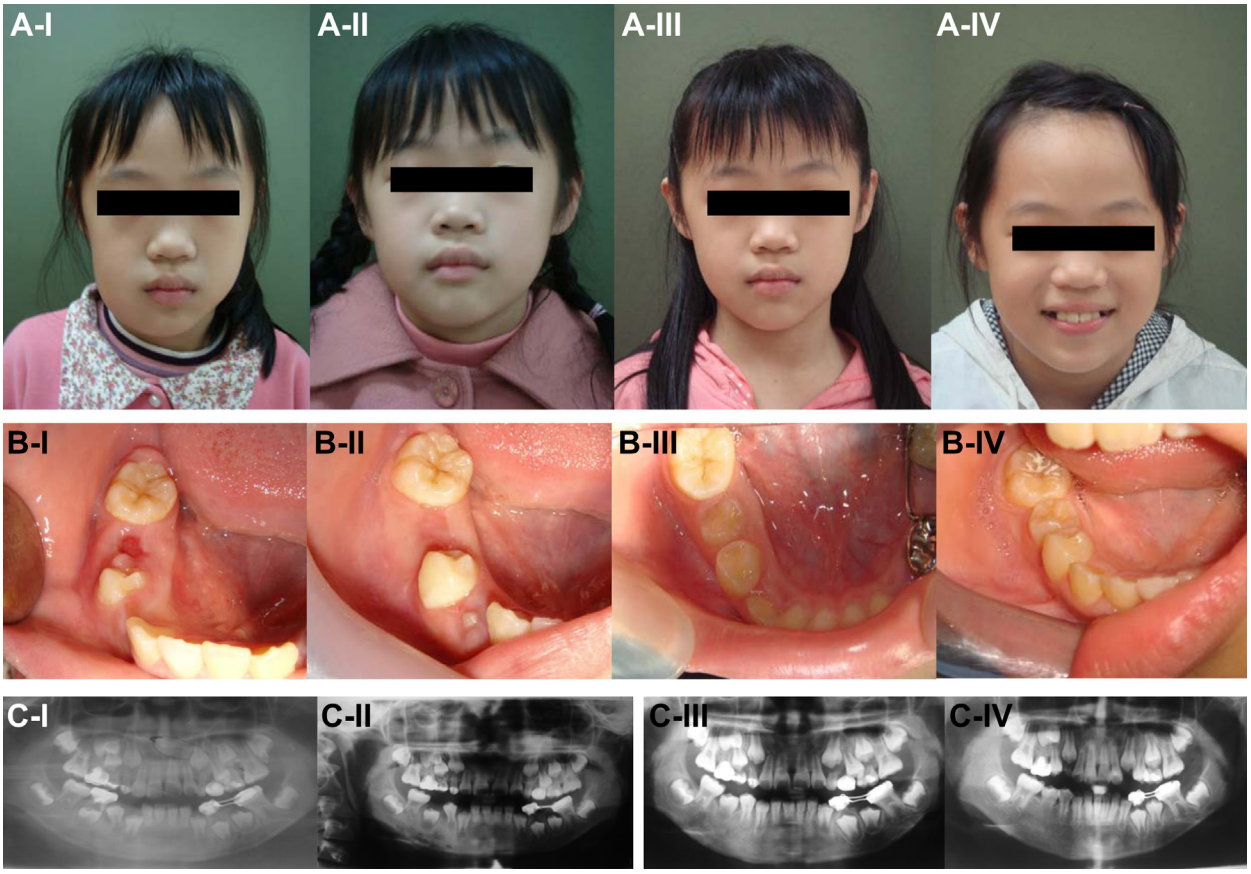

Figure 3 After 10 months, on a recall visit we found the patient to be asymptomatic. (A) Extra-oral photograph. (B) Intra-oral photographs. I: at the first visit; II: at the second visit after a definite diagnosis (25 days later); III: at the 5-month follow-up; and IV: at the 10-month follow-up. (C) The panoramic views (from 3C-I to 3C-IV). I: 1 week before the first visit; II: at the first visit; III: at the 5-month follow-up; IV: at the 10-month follow-up.

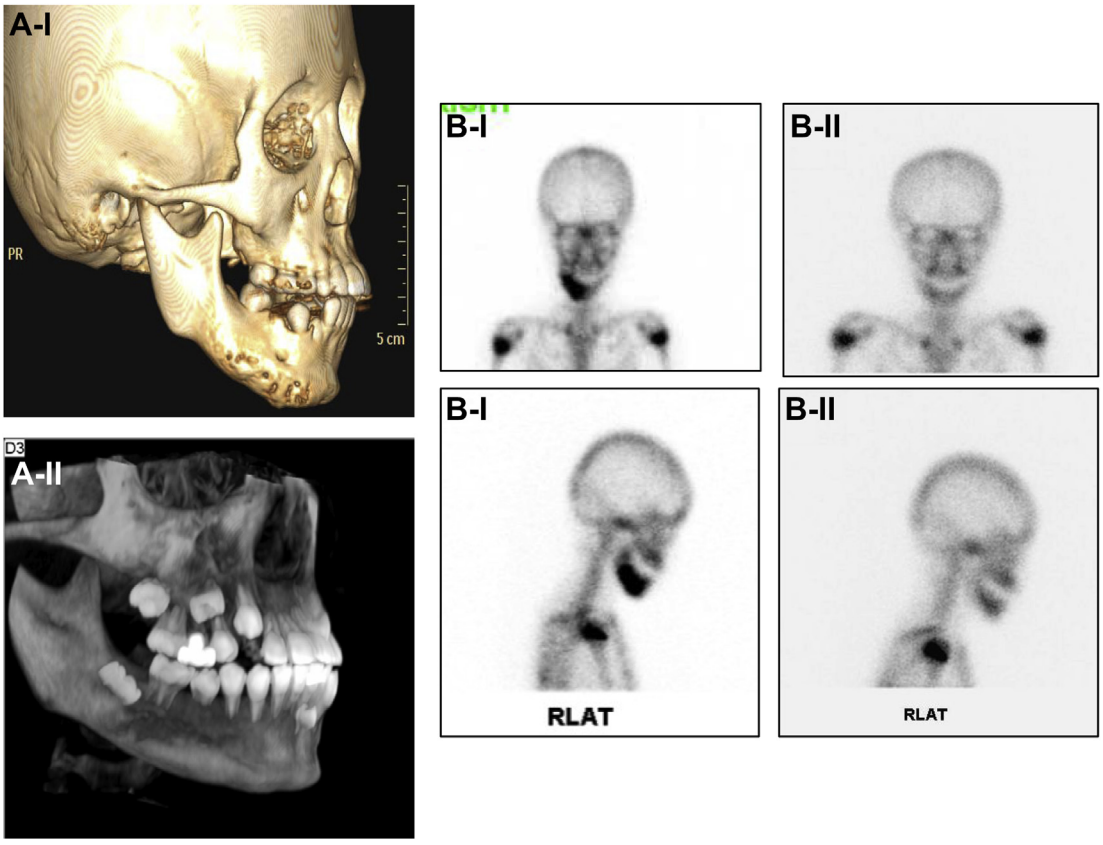

Figure 4 After 10 months, on a recall visit, a computed tomography scan demonstrated remodeling of the mandible, and the region that had shown increased tracer uptake had returned to normal on bone scanning. (A) Three-dimensional computed tomography imaging. (B) Whole-body bone scan images. I: at the first visit; II: at the 10-month follow-up.

CT has made it possible to observe lesions in slices of a few millimeters thickness and three-dimensional images can be constructed from those slices. Therefore, CT has been widely used for diagnosing maxillofacial diseases. In this case, we used CT scanning to investigate the lesion in greater detail, and this played an important role in the diagnosis and monitoring. We used plain and three-dimensional CT to evaluate the status of the lesion and the origin of inflammation. These images showed severe enhancement over the periosteum (Fig. 1B) and an additional, irregular region of cortical bone extending from the parasymphysis to the right retromolar area (Fig. 1C).

Alkaline phosphatase (ALP) is present in all tissues throughout the body, but is particularly concentrated in the liver, bile duct, kidney, bone, and the placenta. The normal range is 20—140 U/L. Elevated ALP indicates that there could be active bone formation occurring as ALP is a by-product of osteoblast activity. The ALP level of this patient (221 U/L) was consistent with the clinical and radiographic examination and initial diagnosis.

The treatment of patients with COPP aims to remove identifiable sources of inflammation. Usually, removal of the infected tooth and curettage of its socket are required to cure the lesion. In some cases, however, endodontic treatment has been reported as a successful means of treating odontogenic causes of proliferative periostitis. As the patient's infected tooth had been removed by a general practitioner and she had been treated for an assumed dentoalveolar abscess with various courses of antibiotics, the identifiable source of inflammation had also been removed. In such cases, conservative treatment should be performed continuously. Treatment includes symptomatic control with non-steroidal anti-inflammatory medication. Remission of disease process and reappearance of a normal-looking mandible was observed by CT imaging, a three-dimensional reconstruction (Fig. 4A) and a bone scan at 10-months' follow-up (Fig. 4B).

This case provides a summary of the lesions included in the differential diagnosis of chronic osteomyelitis with proliferative periostitis and its typical clinical and radiographic features.

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